Multiple supernumerary teeth not associated with complex syndromes: A retrospective study

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Abstract
Objectives: To determine the epidemiology and describe the clinical and radiographic characteristics, the type of treatment, and the possible delayed appearance of new supernumerary teeth in patients with non-syndromic multiple hyperdontia.

Patients and Methods: We conducted a small retrospective observational study of 8 patients diagnosed with non-syndromic multiple hyperdontia. Multiple hyperdontia not associated to complex syndromes was defined as apparently generally healthy patients with one or more supernumerary teeth in two or more areas.

Results: The average patient age was 16.23 years; males predominated (3:1). Multiple hyperdontia with a minimum of 2 and a maximum of 9 supernumerary teeth was found (total: 34 mean: 4.25). The most frequent location was the upper jaw (76.47%). Eumorphic teeth were seen at lower premolar level, while the rest were all heteromorphic. There was altered eruption of the contiguous teeth of 4 of the impacted supernumerary teeth all the rest being asymptomatic. Extraction was the treatment in all patients, and in one of them the delayed appearance of 4 supernumerary teeth was detected.

Conclusions: Multiple hyperdontia rarely occurs without being associated with complex syndromes. Prophylactic surgical removal of the supernumerary teeth is generally the treatment of choice.

Key words: Multiple hyperdontia, multiple supernumerary teeth, non-syndromic hyperdontia.
Introduction
Supernumerary teeth, or hyperdontia, is defined as teeth that exceed the normal dental formula, regardless of their location and morphology (1-3). Such teeth are relatively common, as reflected by the many studies published in the literature, describing this pathology and establishing a prevalence of supernumerary teeth in the permanent and temporary dentition of 0.5-5.3% and 0.2-0.8%, respectively. In addition, the disorder is more common in males than in females, with a proportion of 2:1 (3-5).

Hyperdontia is considered to be multiple when there are one or more supernumerary teeth in two or more dental groups (6). Such hyperdontia is often associated with Gardner syndrome, Fabry-Anderson syndrome, Ehler-Danlos syndrome, facial fissures or cleidocranial dysplasia (1, 6, 7). In contrast, hyperdontia in the absence of such complex syndromes is rare. This type of hyperdontia appears to occur only in the permanent dentition, and usually involves the coexistence of supernumerary teeth in the anterior sector and premolar region – though there have been reports of cases involving all dental series in both maxillae (2, 6, 8).

The etiology is unknown, although a number of theories have been proposed: atavism, tooth germ dichotomy, hyperactivity of the dental lamina, and genetic factors comprising a dominant autosomal trait characterized by low penetrance (6, 8).

Such teeth can erupt normally, remain impacted, invert, reach heterotopic positions, or show abnormal eruptive patterns (9). Likewise, the development of supernumerary teeth can give rise to a broad range of complications, such as delayed eruption of permanent teeth, diastemas, the rotation of adjacent teeth, and cyst lesions or the reabsorption of contiguous teeth. Therefore, an early diagnosis and adequate treatment are essential (3, 4, 10, 11).

The present study was designed to determine the epidemiology and describe the clinical and radiographic characteristics, the type of treatment, and the possible delayed appearance of new supernumerary teeth in patients with non-syndromic multiple hyperdontia diagnosed in a Service of Oral Surgery. An updated review is also made of the literature on the subject.

Patients and Methods
A retrospective observational study was made of all the patients with supernumerary teeth seen in the Service of Oral Surgery of the University of Barcelona Dental Clinic (Barcelona, Spain) between January 2000 and December 2006. Of the 16,384 studied clinical histories, we found 52 patients with hiperodontia, and only 8 presented a multiple hyperdontia not associated to complex syndromes.

The patient data were collected from the clinical histories of the patients, including their panoramic X-rays. Multiple hyperdontia not associated to complex syndromes was defined as healthy patients with one or more supernumerary teeth in two or more dental groups, in the absence of any associated systemic clinical syndrome.

After identification of the cases, the following data were collected: patient age and sex, number of supernumerary teeth per patient, location and morphology of the supernumerary teeth, their clinical manifestations, the management approach used, and the delayed appearance of new supernumerary teeth.

Results
A total of 8 patients (6 males and 2 females) presented multiple hyperdontia not associated to complex syndromes, showing a prevalence of 0.049%. The mean patient age was 16.23 years (range 11-25 years). The presentations ranged from a minimum of 2 supernumerary teeth to a maximum of 9. The total number of supernumerary teeth was 34, with an average of 4.25 per patient.

Most of the supernumerary teeth were located in the upper jaw 76.47% (Fig. 1), with the remaining 23.53% in the mandible. The mesiodens was the most frequent supernumerary tooth (representing 35.29% of the total supernumerary teeth), followed by the supernumerary upper molars (32.35%), the supernumerary lower premolars (23.53%), and the supernumerary upper premolars (8.83%). There were no supernumerary teeth in the rest of dental series.

Of the global 34 supernumerary teeth, 29 were found to be impacted (85.29%), while 2 upper mesiodens, 1 supernumerary lower premolar and 2 supernumerary upper premolars were fully or partially erupted. There were 8 eumorphic teeth – all developing in the lower premolar zone. The rest of the supernumerary teeth were all heteromorphic.

One patient showed altered eruption of 2 upper second molars associated to the presence of 2 erupted paramolars, while in another patient the 2 upper third molars were impacted as a result of 2 impacted paramolars (Fig. 2). The rest of the supernumerary teeth were asymptomatic and caused no complications of any kind.

Treatment in all cases consisted of surgical removal of the supernumerary teeth, followed by clinical and radiographic follow-up to detect the possible delayed appearance of new supernumerary teeth. In this regard, one patient showed 4 supernumerary teeth three years after first being seen in the Service (Fig. 3).

Discussion
There are few published cases of multiple supernumerary teeth not associated to complex syndromes. We thus consider the present series of 8 such cases to be of in-
Multiple supernumerary teeth

Fig. 1. Multiple hyperdontia with the presence of 5 supernumerary teeth. Panoramic X-ray view showing several supernumerary teeth affecting only the upper jaw: one mesiodens, one supernumerary upper left premolar, and three supernumerary teeth in the upper left and right molar groups.

Fig. 2. Panoramic X-ray view showing three supernumerary teeth: two paramolars adjacent to the upper right and left third molars, and one supernumerary tooth between the mandibular right canine and first premolar. The upper paramolars are seen to alter eruption of the upper third molars.

Fig. 3. Clinical case of a male developing a total of 9 supernumerary teeth. A) Initial panoramic X-ray view of the patient at 12 years of age. Five supernumerary teeth are observed: two supernumerary upper incisors, two paramolars at the level of the maxillary right and left second molars, and one supernumerary tooth between the mandibular right canine and first premolar. B) Control panoramic X-ray view 10 months after removal of the supernumerary upper incisors and of the paramolar associated to the upper right second molar. Also a conducting alveolectomy of this tooth was made. C) Panoramic X-ray view 12 months after surgical removal of the supernumerary lower premolar located between the mandibular right canine and first premolar, and of the paramolar associated to the maxillary left second molar. Posteriorly, a conducting alveolectomy of the upper left second molar was performed. D) Late appearance of 4 supernumerary teeth: Two upper distomolars, one supernumerary lower premolar at the level of the mandibular right second premolar, and a supernumerary upper premolar between the upper right premolars.
terest. These were healthy patients with no mental retardation; their facial appearance was normal; and they presented no skeletal or other abnormalities suggestive of a systemic syndrome.

Different studies have reported a prevalence of supernumerary teeth in the permanent dentition of between 0.15% and 3.8% (11, 12). Supernumerary teeth are single teeth in 76-86% of cases, while two supernumerary teeth are found in 12-23% of cases, and three or more such teeth in the same individual are only found in 2-8% of cases (4, 5). However, according to Rajab and Hamdan (4) this percentage is less than 1% when hyperdontia comprises 5 or more supernumerary disease, while Açıkgoz et al. (11) report a 0.06% prevalence of multiple supernumerary teeth. Despite the rarity of such cases, the present study comprises 8 patients with multiple hyperdontia not associated to complex syndromes. An average of 4.25 supernumerary teeth per patient was observed, and two of the subjects had 5 or more supernumerary teeth.

In 1990, Yusof (13) presented a review of the majority of cases of multiple hyperdontia published in the English-speaking literature between 1969 and 1990, concluding that the condition is more common in males (9:2 over females). Our own findings coincide with the observations published to date, since 6 of the 8 patients were males.

The etiology of hyperdontia is not known, though a number of theories have been proposed. The most commonly accepted explanations are dichotomy and hyperactivity of the dental lamina (4, 6, 13). On the other hand, genetic factors appear to be strongly implicated in the origin this pathology. Although until recently the precise mechanism of transmission was not known, the underlying hereditary pattern has gradually been elucidated, thanks to the publication of different cases in which several members of one same family presented hyperdontia (2). Although multiple supernumerary teeth appear to develop more often in patients with some relative with at least one supernumerary tooth, the hereditary trait does not exhibit a simple Mendelian pattern (14). A review of the literature yielded 7 families in which at least two individuals presented multiple supernumerary teeth — with no associated complex syndrome (3, 14-19). Although few such cases have been published, it is important to stress that an exhaustive and detailed family history must be compiled when dealing with patients with multiple hyperdontia, since the dominant autosomal hereditary trait involved implies that all generations are affected. However, although the literature points to a familial predisposition to hyperdontia, in our series there were no supernumerary teeth in any of the direct relatives of the patients. This was probably due to the low penetrance of dominant autosomal transmission, which implies that some generations are not affected by the disorder.

A review of the literature yielded several studies in which large series of supernumerary teeth were analyzed (4, 5, 11). In the case of simple hyperdontia, the supernumerary teeth were most often located in the maxilla — the mesiodens being the most common supernumerary tooth, followed by supernumerary premolars and supernumerary fourth molars (4, 5, 10-12). In contrast, on examining the few published cases of multiple hyperdontia not associated to complex syndromes, a predilection for the mandible and premolar series was observed (13). In our series, and coinciding with a number of authors (1, 3, 8, 20) there were more supernumerary teeth in the maxilla than in the mandible. However, other studies (11, 14, 21, 22) report a predilection for the mandible, in coincidence with the observations published by Yusof (13) in 1990. On the other hand, as regards distribution among the different dental series, most authors have reported an increased presence of supernumerary teeth in the premolar series (1, 3, 13, 14, 19, 21, 22). However, in our study, and in agreement with other investigators (2, 8), there were more supernumerary teeth in the premaxillary zone (twelve); followed in descending order of frequency by the upper molars (eleven), and the lower premolar sector (eight). As an exceptional finding, Desai and Shah (3) presented a case in which all possible supernumerary locations were involved, i.e., affecting all the dental series.

Supernumerary teeth may erupt or remain impacted within the maxilla. Approximately 75% of all supernumerary teeth are impacted and are asymptomatic. Consequently, most such teeth constitute casual findings in the context of routine X-ray studies (4, 5, 11). Our data coincide with those published in the literature, since the great majority of supernumerary teeth (85.29%) were found to be impacted and were identified by chance on performing a panoramic X-ray study for other reasons. It has been reported that supernumerary teeth may be associated to other dental anomalies such as hypodontia, taurodontism, gemination or macrodontia (23, 24). However, no such anomalies were observed in our series, in coincidence with the findings of Baccetti (25), who recorded no relationship between supernumerary teeth and other dental anomalies. Accordingly, this author was of the opinion that supernumerary teeth should be regarded as a distinct pathological entity.

Regardless of whether they are erupted or impacted, supernumerary teeth can induce or be associated with different disorders. Thus, for example, the following has been described in relation to these teeth: delayed eruption or non-eruption of permanent teeth, displacement or rotation of the latter, absorption of roots or of adjacent teeth, root malformations secondary to the pressure exerted by the supernumerary teeth, and the development of cysts (4, 6, 10, 11, 26, 27). Of the above, delayed eruption or non-eruption of permanent teeth, and malformation of
the neighboring teeth are the most commonly reported alterations (27, 28). Nazif et al. (29) reported a 30% prevalence of complications secondary to, or associated with, supernumerary teeth - while Açikgöz et al. (11) reported a prevalence of 21.6%. In our series, eruptive alterations of four upper molars were seen to occur due to the presence of four paramolars – this representing a complications rate of 11.76%. In contrast to the general opinion that cysts are only rarely formed from a supernumerary tooth, Hopcraft (30) found such lesions in 9% of all cases. In our series, however, there were no cysts or enlarged dental follicles. The literature describes different management options for patients with multiple hyperdontia not associated to complex syndromes. Treatment is partly dependent upon the position and clinical manifestations of the supernumerary tooth. Thus, an early diagnosis is very important in order to decide among extraction, extraction followed by orthodontic treatment, or simply monitoring or control of the supernumerary teeth, with a view to minimizing the risk of complications secondary to the presence of these teeth.

Surgical management in turn ranges from removal of the supernumerary teeth to removal of the latter followed by orthodontic treatment aiming to ensure correct occlusion. In the more complex cases, the possible existence of multiple impactions of supernumerary teeth gives rise to destructuring of the dental arch, with numerous malpositioned teeth. These situations require close cooperation among professionals to define combined surgical-orthodontic management. In such cases the orthodontist defines the general management lines, specifying which teeth must be removed or preserved in order to optimize occlusion through orthodontic treatment (6, 18, 26, 31). However, these complicated multiple impactions are almost exclusively found in patients with complex syndromes (7, 18). In our series orthodontic treatment was not needed in any case, since all patients had a normal number of permanent teeth, and occlusion was moreover correct. As a result, treatment was limited to surgical extraction of the supernumerary teeth. If such teeth cause delayed eruption, tooth displacements, root reabsorption or cysts, etc., extraction should be performed as soon as possible (2, 6), since in clinical practice the complications of early extraction are infrequent or only minor (20). In our series there were only four cases of impacted permanent teeth, requiring extraction of the implicated supernumerary teeth. Nevertheless, and in contrast to the approach of other authors (1, 21, 22, 30, 31), prophylactic removal of all the other supernumerary teeth was decided in order to prevent possible complications, since such teeth when left in place can erupt, alter occlusion, or give rise to other disorders (5, 6, 10, 11, 26). Lastly, surgical intervention in our patients did not give rise to complications. Since there is always a risk of damaging some adjacent anatomical structure during extraction, the risk-benefit ratio of tooth removal must be evaluated in all cases (20, 30, 31). As has been described by most authors, the current tendency is not to extract asymptomatic supernumerary teeth, with a view to avoiding possible postoperative complications. In this sense, Hopcraft (30) suggested that particularly in the case of supernumerary lower premolars, the latter must be monitored – since extraction entails a high risk of loss of vitality or root damage of the adjacent teeth. In comparison, the incidence of pathological problems resulting from these impacted teeth is relatively low (11, 22, 30, 32). This is why periodic clinical and radiographic controls are advised, since pathological changes may sometimes occur over the long term (6, 10, 31). Follow-up is necessary, since it is impossible to predict when a follicular cyst will develop, or when reabsorption of a tooth adjacent to an impacted tooth may occur (6). Furthermore, since new supernumerary teeth may appear over time, extraction remains as a treatment option to be indicated or not, depending on the clinical and radiographic findings of posterior controls (1, 10, 11).

Lastly, mention should be made of the possibility of delayed development of supernumerary teeth. A first consideration is that supernumerary teeth begin to develop later than the teeth of the dental series in which they appear. As a result, a common finding is incomplete root formation of the supernumerary tooth, in contrast to full development of the roots of the normal teeth. For this reason, and because most supernumerary teeth lie palatal / lingual, it is often difficult to determine exactly when a supernumerary tooth begins to form on the basis of the routine X-ray findings. As a result, such teeth may sometimes go unnoticed (31). The literature contains a number of cases of late developing supernumerary teeth (1, 2, 8, 31). As an example, Cochrane et al. (31) reported a case in which two late supernumerary lower molars appeared, along with another case in which two lower premolars appeared. Trull et al. (8) in turn published a case in which a late supernumerary lower premolar and upper molar were observed. After reviewing the literature, it is concluded that these teeth normally form in the premolar and molar regions. In our series, and coinciding with the literature, there were four late supernumerary teeth: one upper premolar, one lower premolar, and two upper molars.

Conclusions
Multiple hyperdontia is rarely not associated to complex syndromes. The condition is infrequent and is normally asymptomatic. The diagnosis is usually established as a result of a casual finding when performing routine panoramic X-ray studies. Prophylactic surgical removal of the supernumerary teeth, with resolution of their com-
applications, is generally the treatment of choice. Lastly, emphasis is placed on the need to perform periodic radiographic follow-up, due to the possible late appearance of new supernumerary teeth.

References